

Incidental finding of bilateral papillary thyroid carcinoma in a patient with primary hyperparathyroidism

Abstract

Primary hyperparathyroidism (PHP) affects 0.5%-1% of the adult population and presents with classical signs of renal lithiasis, cholecystolithiasis, gastrointestinal ulcerations, depression, and osteoporosis. Parathyroid adenoma, hyperplasia and rarely carcinoma are the underlying pathology. Synchronous thyroid and parathyroid pathologies are described in multiple endocrine neoplasia. We report a case of a 47 years old woman with non-syndromic concomitant occurrence of bilateral non-medullary thyroid carcinoma diagnosed by histopathology, and with PHP confirmed by ^{99m}Tc -MIBI scintigraphy, hypercalcemia and elevated serum parathyroid hormone. A head and neck surgeon needs to be aware of the possible coexistence of thyroid and parathyroid lesions. To our knowledge, this is the first report of concomitant PHP and bilateral papillary thyroid cancer in the literature. *In conclusion*, it is optimal to remove both tumors in one operative procedure. Therefore careful thyroid evaluation should be considered for all patients with PHP.

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Introduction

The pathological association of thyroid and parathyroid disease is a common finding and is reported in 15% to 70% of patients with primary hyperparathyroidism (PHP) [1-7]. Both malignant and benign thyroid lesions such as in Hashimoto's thyroiditis, goiter with nodular hyperplasia, medullary carcinoma and papillary carcinoma may be found [6, 8]. The thyroid is traditionally evaluated intraoperatively by gross inspection and palpation [6]. However, nowadays, the preoperative workups such as scintigraphic localization of hyperfunctioning parathyroid tissue and high-resolution ultrasound are recommended for the exact localization of lesions.

By review of the literature from 1956 until now, the incidence of thyroid cancer in patients with PHP was reported to be 1.7% to 6% and the cancer was detected unilateral [6, 8]. Our case is the first report of synchronous PHP and bilateral papillary thyroid carcinoma.

Description of the case

A 47 years old woman referred to our hospital with nephrolithiasis and recurrent renal colici. There was no history of familial hyperparathyroidism or multiple endocrine neoplasia or of previous neck irradiation. Biochemical laboratory tests revealed persistent hypercalcemia, hypophosphatemia, elevated urine calcium and elevated serum parathyroid hormone (PTH), (PTH: 88pg/ml; normal range: 10-60pg/ml, serum calcium: 4.2mmol/L; normal range: 2.12-2.55mmol/L). Thus, the diagnosis of hyperparathyroidism was established.

Parathyroid scintigraphy was performed with the intravenous injection of 740MBq ^{99m}Tc -methoxy isobutyl isonitryl (^{99m}Tc -MIBI). Scanning was performed at 15 and 120min and revealed asymmetric thyroid uptake in the early images. Delayed 2h scanning revealed an area of intense ^{99m}Tc -MIBI activity inferior to the left lower thyroid pole that was a well defined parathyroid lesion (Fig. 1A and 1B). Thyroid scan with 5mCi ^{99m}Tc -pertechnetate revealed multinodular goiter with bilateral cold nodules and also a functioning thyroid nodule on the right lobe (Fig. 1C). The patient was operated for both parathyroidectomy and thyroidectomy, therefore, neck exploration was performed. Pathology revealed a parathyroid adenoma of 0.5gr (Fig. 2), a nodule in the right thyroid lobe which contained a tumor of 1.7×1.5cm and another tumor measuring 2×1.5cm in the left thyroid lobe. Pathology study of both tumoral lesions showed papillary structures with fibrovascular core lined with single or stratified cuboidal cells with ground glass nuclei. These findings are typical for papillary carcinoma (Fig. 3A and B). The dissected lymph

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- Primary hyperparathyroidism
- Papillary carcinoma
- Bilateral thyroid carcinoma
- Parathyroid scintigraphy

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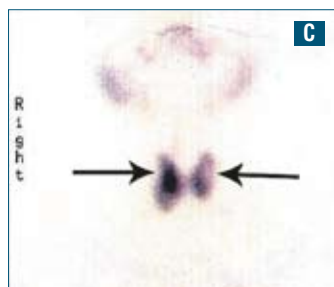
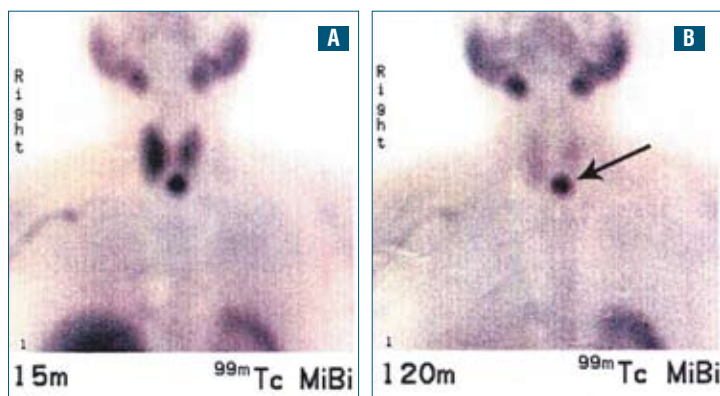


Figure 1A, B and C. Parathyroid scan: A: Fifteen minute ^{99m}Tc-MiBi scan identifies early asymmetric thyroid uptake. B: Delayed 2h scan and also figure A, demonstrate an area of intense ^{99m}Tc-MiBi activity, inferior to the left lower thyroid pole. C: Thyroid scan with ^{99m}Tc-pertechnetate revealed multinodular goiter with bilateral cold nodules and also a functioning nodule on the right lobe.

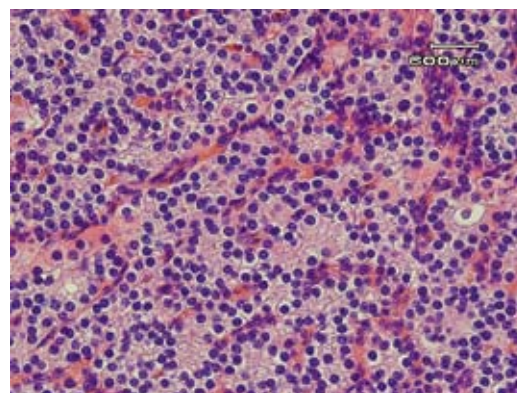


Figure 2. Diffuse proliferation of chief cells with uniform dark nuclei. No stromal fat is present (H&E, magnification x400).

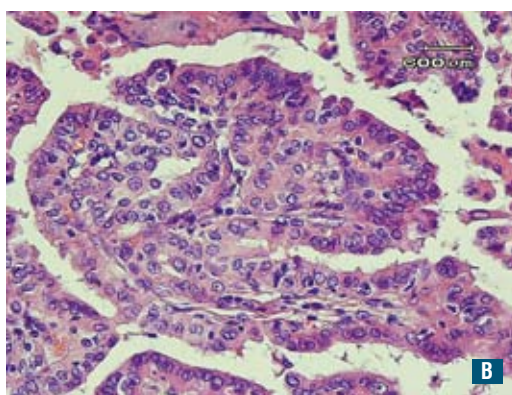
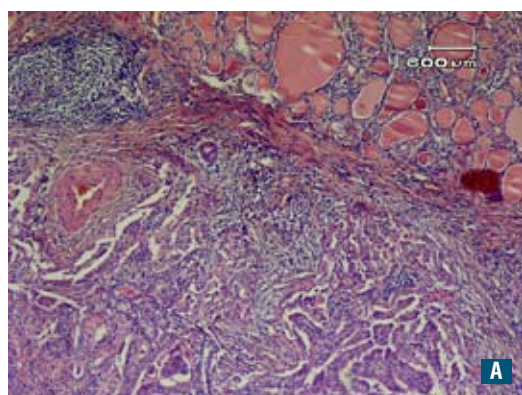


Figure 3A and B: A: Papillary carcinoma adjacent to normal thyroid gland (H&E, magnification x100). B: Papillary structure lined by cuboidal cells with eosinophilic cytoplasm, nuclear clearing and overlapping is shown (H&E, magnification x400).

nodes showed reactive changes. After surgery, PTH and calcium levels returned to normal and remained normal during one year of follow-up.

Discussion

Synchronous thyroid and parathyroid lesions were first described in 1947 [1]. The coexistence of non-medullary thyroid carcinoma and parathyroid adenoma is an uncommon clinical finding. In previously published studies, unilateral thyroid carcinomas were incidentally diagnosed during or after treatment of parathyroid disease, mainly in pathology specimens. Review of the literature indicated that although coexisting benign or malignant thyroid and parathyroid pathology was found in 15% to 70% of patients being treated for PHP [2-6], only approximately 3% of patients operated for PHP had coexisting unilateral non-medullary thyroid cancer [2, 6-8]. By review of the literature, this is the first case report of concomitant PHP and bilateral papillary thyroid cancer.

Previous neck irradiation, especially in childhood, appears

to be a risk factor for the development of both non-medullary thyroid carcinoma and PHP [9, 10]. In medical practice, serum PTH is determined before neck incision and 10-15min after excision of enlarged parathyroid gland(s). A PTH decrease of more than 50% and/or into the normal range are the criteria of successful parathyroidectomy [6, 11-13].

Since coexisting thyroid and parathyroid pathology has been found in many patients, evaluation of the thyroid gland in patients undergoing surgery for parathyroid disease is important [8-12]. During the last decade, preoperative studies such as ^{99m}Tc-MiBi scintigraphy and ultrasound have been used for the evaluation of parathyroid lesions [14, 15]. The ^{99m}Tc-MiBi scan indicates the site of parathyroid hyperactivity, but synchronous thyroid lesions can not be properly evaluated [14]. Thus an extra thyroid scan is indicated.

In conclusion, in order to diagnose coexisting thyroid cancer in patients with hyperparathyroidism, routine thyroid scintigraphy may be applied as shown in our case of bilateral thyroid carcinoma.

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